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Tremor and Other Hyperkinetic Movements

Brief Reports

"Closing the Loop" in Cervical Dystonia: A New Clinical Phenomenon

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Abstract

Background: The sensory trick or geste antagoniste is a cardinal feature of cervical dystonia. Patients are often aware of their tricks, using them to their advantage to temporarily improve dystonic symptoms. The typical sensory trick must be internally generated by the patient in order to be effective, and external mechanical pressure alone may not be sufficient.

Methods: We present a case description. We present and demonstrate a new clinical phenomenon in patients with cervical dystonia, which we call "closing the loop".

Discussion: We discuss the possible mechanisms underlying this finding.

Keywords: Sensory trick, geste antagoniste, cervical dystonia

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Introduction

Sensory tricks, also called "geste antagonistes", are a cardinal feature of many forms of focal dystonia. Gestes usually occur early in the course of the disorder, and loss of efficacy of a geste may signify progression of dystonia.[1] Typical gestes in cervical dystonia (CD) patients include having the patient touch the side of their cheek or chin with their hand, or leaning their head against a wall or chair. A true sensory geste requires only a gentle touch, in contrast to a "forcible" maneuver where considerable pressure must be exerted to normalize neck posture by physically opposing overactive muscles.[2]

The geste antagoniste illustrates the important role of the sensory system in dystonia. For example, vibratory stimuli applied to the hand of a patient with writer's cramp may trigger dystonic movements,[3] and sensory deafferentation of an affected body part by local lidocaine injection may temporarily improve symptoms of dystonia.[4] Features of the clinical examination in CD patients also imply an important role for the sensory system. The pressure needed to obtain benefit from a sensory trick is usually quite mild, far too mild to physically oppose the muscles that are overactive. In many patients, application of the trick does not depend on the direction of the primary dystonic movement. For example, patients with a leftward rotational torticollis may have equal benefit applying gentle touch to either the right or the left side of the chin.[5] Further, many patients experience improvement in dystonia in anticipation of the geste finger contacting its target. Finally, many patients with dystonia who possess prominent sensory tricks can partially reproduce the geste benefit by imagining they are performing the trick (an "interoceptive" trick).[6] These features suggest the possibility that the mechanism of action of sensory tricks may depend on modulation of internal sensorimotor networks. Such a network has been demonstrated in a positron emission tomography study of CD patients, with activation of the left parietal lobule and reduced activation of supplementary motor area during performance of a geste.[7]

The majority of patients with CD only benefit from a sensory trick when they perform it (i.e., passive touch of the chin by the examiner is ineffective). In this paper, we report a new clinical phenomenon in CD, in which having the patient take the examiner's fingers and touch their chin produced an effective sensory trick, while passive touch by the examiner was ineffective. To highlight the possible sensorimotor mechanisms underlying this trick, we call this phenomenon "closing the loop" in CD.

Table 1. Clinical Features of Four Patients with "Closing the Loop" Phenomenon

Patient no.	Age at Onset	Age at Evaluation	Other Dystonia	Severity of CD	Response to Btx	Sensory Tricks	Imagined Geste	Examiner's Touch	"Closing the Loop"
1	42	47	WC	Marked	Poor	Chin, head	–	–	+
2	61	68	–	Moderate	Good	Chin, head	+	–	+
3	56	59	–	Moderate	Good	Chin, head	+	–	+
4	22	27	–	Mild	Good	Chin, head	–	–	+

CD, Cervical Dystonia; Btx, Botulinum Toxin; WC, Writer's Cramp.
Clinical features of four patients are presented: other dystonia refers to the presence of dystonia in other body regions. The examiner rated severity of dystonia as mild, moderate, or marked, and response to Btx injection was similarly rated as poor, partial, or good. Sensory tricks of touching the chin or back of the head benefitted all patients, and two patients experienced benefit from imagining themselves touching their chin. No patients benefitted from the examiner touching their chin at the same location, but all benefitted when the patient put the examiner's hand on the same location ("closing the loop").

Methods

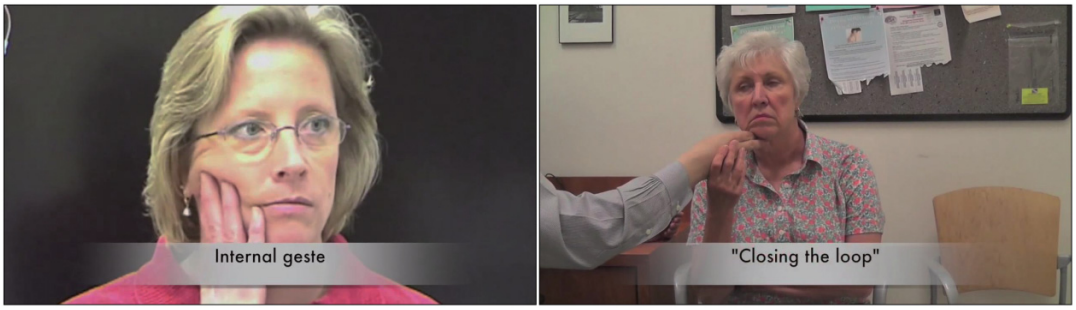
Clinical details of four CD patients encountered in routine clinical practice over 8 years are summarized in Table 1. We looked for the sign in many but not all of the patients with CD examined over this period of time. The examination and phenomenon are demonstrated in two patients in the accompanying videos. All four patients were women with isolated CD, with no history of neuroleptic exposure. One patient also displayed writer's cramp. All patients demonstrated prominent sensory tricks (touching the chin with a hand, or holding the back of the head, or leaning it against a wall). Two of four patients possessed an interoceptive trick, experiencing noticeable improvement by imagining their sensory trick of touching the chin. No patient benefited from an externally applied sensory trick by the examiner (touching the chin at the subject's sensory trick target). All four subjects experienced sensory trick benefit when they held the examiner's fingers and applied them to their chin ("closing the loop"). In all patients, improvement in neck position occurred immediately before the patient performed the trick, and a similar benefit was observed during the "closing the loop" sign (see Video 1).

Discussion

We describe a new phenomenon in CD, "closing the loop". We have chosen this name to highlight the fact that the sensory trick benefit requires a closed tactile circuit in the patient's body, from the patient's chin to the examiner's hand to the patient's hand.

Several groups have investigated the phenomenology and mechanisms underlying the sensory trick in CD. Wissel et al.[8] studied 25 patients using surface electromyography (EMG) to measure the timing of improvement in dystonia relative to contact of the finger with the geste target. Latency of the initiation of the trick and change in surface EMG signal varied from the onset of movement to 0.9 s, and two-thirds of patients did not experience improvement when the examiner touched the geste target site. Muller et al.[9] studied 50 CD patients, observing a reduction in dystonia prior to contact with geste target in one third. Thirty-eight percent of patients experienced at least some benefit from the examiner replicating the trick, but the remainder did not. Ocudlo et al.[10] examined 33 CD patients, including 19 with a prominent geste. Only 32% of these patients experienced a clear improvement in dystonia with the examiner's touch. Taken together, these studies suggest that as many as two-thirds of CD patients experience little to no benefit from an externally applied sensory trick, suggesting that the internal gesture is critical to trick efficacy. It is unknown how many of these patients have a "closing the loop" phenomenon, but we suspect many do.

The existence of imagined sensory gestes and the anticipatory benefit before contact of the hand in certain gestes implies that some patients may benefit substantially from modulation of frontoparietal sensorimotor networks. The "closing the loop" phenomenon is a similar entity.



Video **1. Clinical Examination Findings in Patients 1 and 2.** Patient 1 demonstrated a significant rightward turning torticollis, with prominent improvement with sensory tricks of either touching the right cheek or leaning the back of the head against a wall. Improvement occurred about a half-second before each trick was applied. When the examiner applied gentle pressure to the back of the head or to the side of the face, no benefit was seen. When the patient placed the examiner's fingers on the side of her chin, sensory trick benefit occurred. The second patient illustrates a jerky, leftward turning torticollis with mild right tilt. Prominent sensory trick benefit from touching the chin was not replicated by passive touch of the examiner, but "closing the loop" produced benefit.

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